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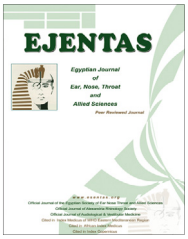

CASE REPORT

Granular cell ameloblastoma : A case report with a brief note on review of literature

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Abstract Ameloblastomas are tumours of odontogenic epithelial origin with varied microscopic patterns that occur either singly or in combination. Granular cell pattern is rarely seen in ameloblastoma, and is characterised by nests of large eosinophilic granular cells. This article describes a case of granular cell ameloblastoma in a 29 year old male patient with clinical, radiology and histological findings along with a short on review of literature.

1. Introduction

Odontogenic tumours are lesions derived from the epithelial or mesenchymal remnants of the tooth forming apparatus.[1] Ameloblastoma, the tumour that meets today's diagnostic criteria for solid/multicystic ameloblastoma (SMA) has been known for about 180 years.[2] The ameloblastoma is a benign but locally invasive tumour and accounts for about 11% of all odontogenic tumours in Caucasians.[3] Granular cell ameloblastoma is a rare variant of ameloblastoma that histopathologically has numerous large granular cells. These usually form the central mass of epithelial islands and cords.[4] In this case report we have presented a rare histological variant of ameloblastoma: Granular cell ameloblastoma.

2. Case report

A 29-year old patient reported to outdoor patient department with a history of swelling in the lower 1/3rd of the face in the front region since two months. A swelling appeared insidiously in the lower left back tooth region two years back about the size of peanut and thereafter gradually it has increased to the present size approximately (10 × 5 cm) and has increased rapidly in the last months. No associated pain, no facial paralysis, no paraesthesia or anaesthesia and no palpable regional lymphadenopathy were present.

Extra oral examination revealed the facial asymmetry in the lower 1/3rd of face (Fig. 1). The swelling was present in the anterior mandible and extended about 3 cm behind the angle of the mouth on both sides. Swelling was approximately 8 × 5 cm in size with smooth surface and colour of skin over the swelling was normal. On palpation, a firm mass was felt

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Figure 1 Clinical photograph showing diffuse swelling in the lower 1/3rd of face.

except parasymphysis where it was hard and tender. Intra orally, swelling was extending from right mandibular 1st pre molar to left mandibular 1st molar and obliteration of the buccal and labial vestibule was noted in the involved site. (Fig. 2) Buccally swelling was firm to hard and slightly tender, lingually swelling was bony hard, non tender and no secondary changes were noted.

Routine biochemical and haematological investigations were within normal limits. Panoramic view revealed the multilocular radiolucent lesion extending from distal root of mandibular right 2nd molar to left mandible left 2nd molar mesial root. (Fig. 3) Following this the incisional biopsy was planned for the patient and histopathology report showed the sections exhibiting the odontogenic epithelium present in the form of islands, cords and few follicles in a connective tissue stroma. Odontogenic islands showed peripherally arranged tall columnar cells surrounding the granular cells (Fig. 4). Granular cells exhibited abundant cytoplasm filled with eosinophilic granules (Fig. 5). Features were suggestive of Granular cell ameloblastoma.

The case was managed by intra oral surgical removal of the tumour mass under general anaesthesia. Post operatively the healing was uneventful and no recurrence was reported.

3. Discussion

The ameloblastoma is a benign odontogenic tumour located almost exclusively in the jaws. It has a distinctive microscopic appearance characterised by the presence of peripheral columnar cells with hyperchromatic, reversely polarised nuclei, arranged in a palisaded pattern.[5] Several microscopic subtypes

Figure 2 Intra oral photograph showing buccal and labial cortical plate expansion and obliteration of the vestibule.

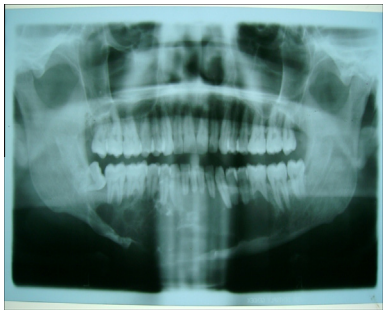

Figure 3 OPG showing multilocular radiolucent lesion extending from distal root of mandibular right 2nd molar to left mandible left 2nd molar mesial root.

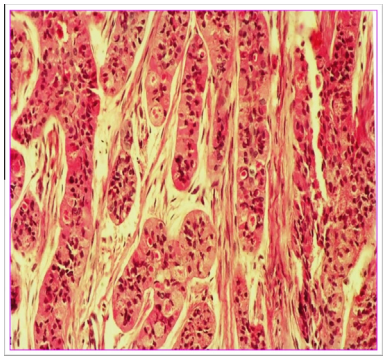

Figure 4 Photomicrograph showing stand and cords of odontogenic epithelium exhibiting granular cells in the centre: (Under 20X).

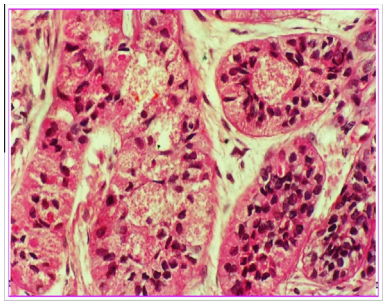

Figure 5 Photomicrograph of granular ameloblastoma under high power showing darkly stained eosinophilic granules in the centre of odontogenic epithelial islands or follicles: (Under 40X).

of the ameloblastoma, especially of its solid/multicystic variant, are recognised, although these microscopic patterns generally have little bearing on the behaviour of tumour. Large tumours often show a combination of microscopic patterns. The follicular and plexiform patterns are the most frequent. Less common histopathologic subtypes include the acanthomatous, granular cell, desmoplastic, and basal cell.[6] Although the treatment and prognosis are virtually the same (with the possible exception of more aggressive desmoplastic variant), knowledge of various histopathologic subtypes is a prerequisite for accurate diagnosis and management.[5]

Granular cell ameloblastoma is a rare variant of ameloblastoma which accounts for only 5% of all ameloblastomas as

stated by Hartman.[7] This term is used when the tumour, most often of the follicular type, shows an extensive granular transformation of the central stellate reticulum like cells. In some lesions all cells of the tumour islands or nests are composed of granular cells.[2] The granular cells may be cuboidal, columnar or rounded and the cytoplasm is filled with acidophilic granules. Ultrastructurally, these granules represent lysosomal aggregates.[8]

In the clinicopathologic study of Kameyama et al. only 1 out of 77 ameloblastoma cases was classified as the granular cell subtype.[9] Reichart et al. reviewed all available literature on ameloblastoma of the jaws from 1960 to 1993 and reported that out of a total of 1593 cases with available data on histologic subtypes, there were only 56 (3.5%) cases of the granular cell variant.[10]

The age distribution of granular cell variant is similar to the other types of ameloblastomas which shows an approximately equal prevalence in the third to seventh decade of life.[5] About 85% of tumours occurred in the mandible, the vast majority of which affected the molar–ramus region.[5] Jaw swelling and pain were the most frequent presenting symptoms. Compared to the other ameloblastoma subtypes, no distinguishing radiographic findings have been reported.

Kumamato and Ooya also performed IHC studies on six cases of granular cell ameloblastomas and demonstrated that the granularity might be caused by increased apoptotic cell death and associated phagocytosis by neighbouring neoplastic cells.[11]

The granular cell ameloblastoma is S-100 protein and alpha-1-anti-trypsin positive. They are negative for vimentin, NSE (Neuro specific enolase), EMA (Epithelial membrane antigen), CMA (Common muscle actin) and Prekeratin.[12]

The biological behaviour of granular cell ameloblastoma does not seem to differ from the other histologic subtypes of ameloblastoma; it can be locally aggressive and has a relatively high chance of recurrence.[13] Reichart et al. reported a 33.3% recurrence rate for granular cell ameloblastoma, which was higher, compared to the more common follicular, plexiform and acanthomatous subtypes.[10] In Hartman's study, 11 of 15 patients (73%) developed recurrent lesions. However, similar to the other types of solid or multicystic ameloblastoma, the prognosis is more dependent on the method of surgical treatment, i.e. granular cell ameloblastomas treated by enucleation or curettage exhibit a high recurrence rate due to the fact that the border of the tumour within cancellous bone lies beyond the apparent macroscopic surface and the radiographic boundaries of the lesion. Therefore radical surgical methods are recommended.[13] Granular cell ameloblastomas may rarely behave in a malignant fashion giving rise to metastasis.[14]

The differential diagnosis of granular cell ameloblastomas includes other oral lesions with a similar morphology of granular cell accumulation, including granular cell odontogenic tumour, granular cell tumour and congenital epulis. These lesions have different biologic behaviours and should be discriminated from granular cell ameloblastomas.[12]

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